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A CASE OF LUPUS CYSTITIS DIAGNOSED FROM URINARY SYMPTOMS AS THE SOLE INITIAL COMPLAINT

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Cystitis related with systemic lupus erythematosis (SLE) is termed as lupus cystitis. It is a subtype of SLE in which gastrointestinal and bladder symptoms are prominent and it usually manifests with other symptoms of SLE. We present a case in a 37-year-old woman whose sole complaint was the severe urinary symptoms. A biopsy of the bladder showed inflammation in the submucosa and the finding of vasculitis. An autoimmune disease was suspected from the blood serum sample. A further laboratory study was performed and she was diagnosed with SLE and lupus cystitis. Prompt therapy with corticosteroids resulted in the improvement of the severe bladder symptoms.
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Key words : Lupus cystitis, Systemic lupus erythematosis, Interstitial cystitis, Autoimmune disease

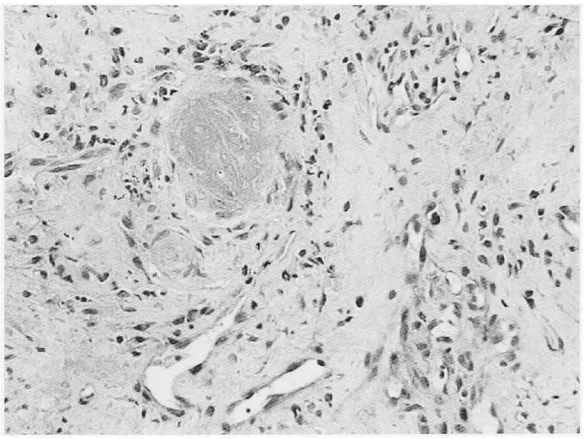
INTRODUCTION

Interstitial cystitis is known to be associated with many autoimmune diseases. In particular, cystitis related with systemic lupus erythematosis (SLE) is termed as lupus cystitis. This type of cystitis is diagnosed in approximately 1% of all the systemic lupus erythematosis cases. It is said to typically present with several symptoms of SLE. When present at the initial diagnosis of SLE, it is usually accompanied by other systemic symptoms such as abdominal symptoms, rash, or arthritis. We present a rare case in which urinary symptoms were the only complaint prior to the diagnosis of SLE and lupus cystitis.

CASE REPORT

The patient is a 37-year-old female who was referred to our urology department. She presented with a week-long history of miction pain, pollakisuria, and gross hematuria. She had been treated with antibiotics for the past week under the diagnosis of cystitis; however, her symptoms showed no sign of improvement. Her past medical history was unremarkable. She had not been on any other medication recently. Her examination revealed that she had severe miction pain, and she was voiding every 15 min. Her bladder capacity was less than 50 ml. A urinalysis showed hematuria and pyuria, although her urine culture and urine cytology tested negative. Her urine also tested negative for tuberculosis. Her physical examination was unremarkable except for a slight malar rash, which was not recognized at the initial outpatient visit.

She had a urethral catheter fitted, which worsened the pain further. An epidural tube was fitted to control the pain. An intravenous pyelography demonstrated a contracted bladder and a normal upper urinary tract. A biopsy of the bladder was performed under cystoscopy. A cystoscopy revealed that her bladder mucosa was diffusely swollen, as is common in the condition of



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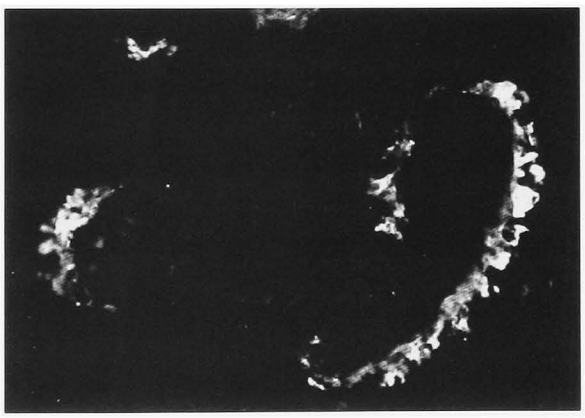

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Fig. 1. A, Hematoxylin and eosin staining of the bladder mucosa shows inflammatory cell aggregation and edema in the submucosa with fibrinoid necrosis of the vessel walls. B, Immunohistochemical staining reveals C3 deposits in the vessel walls. The same pattern was observed with immunohistochemical staining with IgM.

cystitis. No glomerulation was observed. Histological specimen showed inflammatory cell aggregation and edema in the submucosa with fibrinoid necrosis of the vessel walls, and the immunological stain proved IgM and C3 deposits in some vessel walls (Fig. 1). The blood serum sample revealed thrombocytopenia, prolonged activated partial thromboplastin time (APTT), and positive results for the rapid plasma reagin test, which raised the suspicion of an autoimmune disease. Further laboratory investigation proved positive for serum anti-nuclear antibody, anti-ds-DNA antibody, anti-Sm antibody, and anti-cardiolipin antibody. Based on the laboratory findings and a malar rash, the diagnostic criteria for SLE were fulfilled, and she was diagnosed with lupus cystitis.

She was orally administered prednisolone at 50 mg daily and her symptoms temporarily improved. However, one month later, arthritis accompanied with hypocomplementemia emerged. Methylprednisolone pulse therapy suppressed the activity of SLE. She is now maintained with a daily oral dose of 15 mg. A cystoscopy shows a normal bladder mucosa. Her bladder capacity has increased to 300 ml and presently she is free of any urinary symptoms.

DISCUSSION

In 1983, Orth et al. first described lupus cystitis as interstitial cystitis occurring in patients with systemic lupus erythematosis (SLE)[1] It was characterized as the condition having strong association with the presence of gastrointestinal symptoms and showing good response to the corticosteroid therapy. By reviewing the 30 reported cases in Japan, Takabayashi described lupus cystitis as a subtype of SLE in which the gastrointestinal and bladder symptoms are prominent[2] Histologically, it is characterized by inflammatory cell aggregation and edema in the bladder submucosa. In some cases, the immune deposit formation, which is also frequently seen in gastrointestinal mucosa, is seen in the vascular walls. This is suggestive of the association between lupus cystitis and vasculitis due to SLE. No autoimmune antibody specific to this condition has yet been identified. Lupus cystitis has been diagnosed in approximately 1% of SLE patients; however, in autopsy cases, a more frequent association is suggested.

Typically, lupus cystitis patients present with bladder symptoms, such as miction pain and pollakisuria along with abdominal symptoms including nausea, vomiting, and diarrhea. Other symptoms of SLE may also accompany them. The reason for the concurrent occurrence of the abdominal symptoms with the bladder symptoms is unclear; however, the presence of abdominal symptoms in almost all the reported cases denies a mere coincidence[2] Lupus cystitis can be present either at the time of initial diagnosis of SLE or after the diagnosis of SLE. Both the urine culture and urine cytology are negative. Radiologically, hydronephrosis and contracted bladder are often detected by intravenous pyelography, and in some cases, ascites is present. These symptoms are usually reversible with corticosteroid therapy; however, in some cases, due to severe bilateral hydronephrosis, urinary diversion or bladder augmentation is required[3,4] Laboratory data reflecting the activity of SLE, such as anti-nuclear antibody and complements, improve in parallel to the symptoms.

The relationship between interstitial cystitis and autoimmune disorders, including Sjogren's syndrome, inflammatory bowel disease, fibromyalgia, and SLE is recognized; in some cases, the activity of autoimmune disorders run parallel to interstitial cystitis[5] In our case, the histological findings and several laboratory data directly led us to suspect SLE and lupus cystitis. Furthermore, the prompt therapy with corticosteroids resulted in improvement of the severe bladder symptoms. The interstitial cystitis, even in the absence of other systemic symptoms, warrant further examination for autoimmune disorders. Therefore, lupus cystitis should be suspected, particularly when accompanied by nonspecific gastrointestinal symptoms.

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和文抄録

初診時に膀胱刺激症状のみを訴えたループス膀胱炎の1例

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全身性エリテマトーデス（SLE）に伴う膀胱炎はループス膀胱炎と呼ばれる．ループス膀胱炎はSLEの亜型で消化器症状と膀胱症状を有するのが特徴と言われ，通常SLEの他の症状を伴う．症例は37歳，女性．重度の膀胱刺激症状のみを主訴とし受診．膀胱生検で粘膜下の炎症と血管炎の所見を認めた．血液データより自己免疫疾患が疑われたためさらに検査を追加したところSLEおよびループス膀胱炎と診断された．早期のステロイドによる治療により重度の膀胱刺激症状は軽快した．

（泌尿紀要 **51**：685-687，2005）